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Case Report

Stent-graft placement for treatment of massive hemobilia caused by porto-biliary fistula

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ABSTRACT

Proton beam therapy is a type of radiation therapy and a promising modality for cancer management because it involves few adverse effects and high therapeutic efficacy. However, there are reports of acute and late complications because of normal tissue damage. Hemobilia, known as bleeding from the biliary tree, is observed in various conditions, and it can also be of iatrogenic origin such as due to percutaneous hepatobiliary interventions. In most cases, it can be managed conservatively without significant hemorrhage. However, in a few cases with massive hemobilia, further intervention is necessary. We report the successful use of a stent-graft in the portal vein to treat massive hemobilia with porto-biliary fistula that was caused by previous proton beam therapy.

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Keywords: Biliary fistula; Hemobilia; Proton therapy; Self expandable metallic stents

Introduction

Hemobilia is bleeding from the biliary tree and is a condition commonly caused by iatrogenic trauma, accounting for 70% of cases.^{1,2} In most cases, hemobilia resolves spontaneously with conservative management. However, it can be resistant to conservative management and might require embolization via a transarterial approach.³ Additionally, massive hemobilia from a porto-biliary fistula is uncommon, and further intervention is necessary in a few cases.^{3,4}

Proton beam therapy (PBT) is a promising modality for the management of various malignancies. Appropriate application of PBT can lead to fewer adverse effects and higher therapeutic efficacy compared with conventional radiation therapy that uses X-ray beams.⁵ Despite this, there are reports of some adverse reactions including severe complications due to normal tissue damage.^{6,7}

We report a case of massive hemobilia with porto-biliary fistula due to PBT, which was successfully treated by endovascular treatment via a percutaneous transhepatic approach.

Case Report

The Institutional Review Board of Aichi Cancer Center Hospital requires no approval for publication of retrospective case reports.

A 54-year-old male treated with PBT (radiation dose, 76 Gy/20 fractions) for hepatic hilar cholangiocarcinoma referred to our hospital because of recurrence of a tumor (hepatic hilar lymphnode metastasis), which appeared 3 years after undergoing PBT. The tumor was treated by systemic chemotherapy followed by surgical resection. The patient suffered obstructive jaundice 6 years after undergoing PBT due to hilar bile duct stricture. However, pathological examination by the endoscopic retrograde cholangiography (ERC) approach showed no malignancy. Instead, ERC revealed a benign stricture and distal localized saccular dilatation of the hilar bile duct. Consequently, PBT was thought to be a possible cause of these findings (Fig. 1A). Two plastic stents were inserted in the lateral segment of the biliary tree; however, because of stent obstruction, frequent exchange (5 times in 14 months) of plastic stents was needed. To address this, fully covered metallic stents were put in place. Although it led to a less

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Fig. 1. (A) Endoscopic retrograde cholangiography shows the severe stricture and distal localized saccular dilatation of the hilar bile duct (arrow). (B) Contrast-enhanced computed tomography 11 years after proton beam therapy (PBT) reveals blood flow in the saccular dilatation (white arrow) of the hilar bile duct adjacent to the left portal vein (black arrow) and remarkable right hepatic lobe shrinkage due to PBT.

Fig. 2. (A) Percutaneous transhepatic portography performed via the peripheral portal branch at the left lateral superior segment reveals severe stricture of the left portal branch (arrow). (B) Portography shows the extravasation into the duodenum along the outer space of the covered biliary stent (arrowheads). (C) Portography shows severe stricture of the left portal branch with developed cavernous collateral blood flow at the hepatic hilum.

frequent replacement of the biliary stents, massive hemobilia occurred in the fourth exchange of the metallic stents 8 years after PBT. This massive hemobilia was controlled by replacement of the fully covered metallic stents in the biliary tree followed by blood transfusion. A subsequent angiography did not reveal the source of hemobilia. Since then, massive hemobilia recurred during the exchange procedure of the metallic stents. Eleven years after PBT, a contrast-enhanced computed tomography (CT) revealed blood flow in the saccular dilatation of the hilar bile duct adjacent to the left portal vein (Fig. 1B). We further observed a remarkable shrinkage of the right hepatic lobe, a compensated left lobe hypertrophy, and a localized stricture of the left portal branch near the saccular dilatation. Hence, we diagnosed massive hemobilia caused by porto-biliary fistula and began endovascular treatment for hemostasis after informed consent obtained.

Percutaneous transhepatic portography performed via the peripheral portal branch at the left lateral superior segment revealed severe stricture of the left portal branch (Fig. 2A). A 5F catheter could be advanced into the porto-biliary fistula through the stricture using a 0.035-inch guide wire (Radifocus M; Terumo, Tokyo, Japan). Contrast study revealed extravasation of fluids into the duodenum along the outer space of the covered biliary stent (Fig. 2B). Another 5 F catheter was advanced to the main portal trunk

using a guide wire, and portography revealed severe stricture of the left portal branch with cavernous collateral blood flow developed at the hepatic hilum (Fig. 2C). Also, a super stiff guide wire (Amplatz extra-stiff wire; Cook Medical, Bloomington, IN, USA) was inserted into the main portal trunk through the catheter, and an 8 F stent delivery system was passed through the stricture over the guide wire. The stent-graft (10 mm diameter, 60 mm length, Niti-S biliary covered stent; Taewoong Medical, Seoul, Korea) was placed from the main portal trunk to the left portal branch across the porto-biliary fistula (Fig. 3A). Despite post balloon dilation, the stent-graft was strongly bent and not fully expanded at the stricture portion of the portal vein (Fig. 3B, 3C). Immediately after the procedure, antiplatelet therapy was initiated with aspirin at a daily dose of 100 mg (Bayaspirin; Bayer, Leverkusen, Germany). One month after the procedure, contrast-enhanced CT revealed no blood flow in the saccular dilatation portion. However, the stent-graft was occluded by thrombus formation and peripheral portal blood flow could not be preserved (Fig. 3D). During a clinical course after the procedure, there was no hepatic dysfunction despite the stent-graft occlusion. Also, more than 1 year after the stent-graft placement, no further hemobilia was observed during exchange procedure of the biliary metallic stents. Five months after the stent-graft placement, endoscopic cholangioscopy revealed

Fig. 3. (A) The stent-graft is placed from the main portal trunk to the left portal branch across the porto-biliary fistula. (B) Despite a post balloon dilation, the stent-graft is strongly bent and not fully expanded at the stricture portion of the portal vein (arrow). (C) Portography shows recanalization of the left portal branch. However, the stent-graft is not fully expanded (arrow). (D) Contrast-enhanced computed tomography 1 month after the procedure shows no blood flow in the saccular dilatation portion (arrow). However, the stent-graft (arrowhead) is occluded by thrombus formation, and peripheral portal branch flow could not be preserved.

a wall defect at the hilar bile duct that was entirely covered with the portal vein stent-graft.

Discussion

We herein report the successful treatment of massive hemobilia with porto-biliary fistula that occurred 8 years after PBT, using stent-graft placement in the portal vein. Complete hemostasis was achieved, and endoscopic cholangioscopy after the procedure revealed a large hole at the hilar bile duct, which was covered with a stent-graft.

A common bile duct stricture is sometimes encountered as a late complication of radiation therapy, and it causes damage in biliary microcirculation environment with radiation-induced injury of peribiliary arterial endothelium.^{7–9} Although PBT is a promising modality for the management of various malignancies because of the possibility of radiation dose reduction to critical organs, acute and late toxicities due to normal tissue damage have been reported.^{5–7} The incidence of cholangitis and common bile duct stenosis (two late complications) previously reported for PBT in advanced cholangiocarcinoma was 21.4% (6/28) and 3.6% (1/28), respectively.⁷ In our case, we observed the stricture and distal localized saccular dilatation of the hilar bile duct. Tissue damage due to PBT and mechanical irritation from the exchange of biliary metallic stents were thought to be a possible cause of the porto-biliary fistula.

Hemobilia arises where there is communication between a vascular structure and the biliary tree.¹ Although hemobilia is sometimes observed after percutaneous hepatobiliary interventions, it can be managed conservatively in most cases that result in portal injury.^{2,3} However, massive hemobilia due to porto-biliary fistula may require more invasive treatment, such as coil embolization and stent-graft placement in the injured portal vein.^{3,4} We confirmed clearly by portography extravasation from the portal vein to the biliary tree and successfully covered the fistulous portion in the portal vein by stent-graft. Because there were no available covered stents for peripheral veins in our country, a biliary covered stent was used. Coil or glue embolization in the portal vein may be other considerable treatment options for closure of porto-biliary fistula.¹⁰ However, in our case, stent-graft placement was better treatment because the biliary wall defect was so large that coil or glue might migrate into the biliary tree via the porto-biliary fistula. Preservation of portal blood flow was an additional benefit with stent-graft placement compared with

coil or glue embolization. Although preservation of portal blood flow failed due to an unexpected occlusion of the stent-graft, fortunately, no hepatic dysfunction or parenchymal shrinkage was observed because hepatic arterial blood flow was maintained. As is the case with percutaneous coronary intervention, the use of anticoagulants (warfarin sodium) or dual antiplatelet therapy (aspirin and clopidogrel) should be considered to prevent early stent occlusion.¹¹ However, those therapies were not initiated in our case because of risk of rebleeding during further endoscopic procedure. Therefore, aspirin alone was used as antiplatelet therapy in our case.

In conclusion, endovascular treatment was effective for massive hemobilia caused by porto-biliary fistula due to previous PBT. Although PBT is a promising modality for cancer management, we should pay careful attention to the possibility of late complications due to normal tissue damage.

Conflicts of Interest

No potential conflict of interest relevant to this article was reported.

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